



An unusual case report of transcatheter aortic valve implantation-related actinomycosis endocarditis

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Background

Actinomycosis is a chronic invasive infection caused by *Actinomyces* species. Actinomycosis endocarditis has been described, yet considered rare. We present the first reported transcatheter aortic valve implantation (TAVI)-related actinomycosis endocarditis.

Case summary

A 70-year-old female patient, presented 4 months after TAVI with malaise and vocal-cord paralysis. She underwent computed tomography angiography which demonstrated a 28 mm pseudoaneurysm of the ascending aorta, which compressed the laryngeal nerves. Her condition rapidly deteriorated with cardiogenic shock and required an emergent surgery, which revealed a tamponade with active bleeding, due to an ascending aortic dissection. She underwent aortic valve and ascending aorta replacement. A 2 cm vegetation was found on the TAVI prosthetic valve and sent for cultures, which later revealed an *Actinomyces neuui* infection. Long-term intravenous ampicillin treatment was given.

Discussion

This case describes a patient with endocarditis on TAVI prosthetic valve, with an unusual clinical presentation and rapid deterioration to an emergency intervention. This unique presentation of tumour-like tissue invasion is characteristic of actinomycosis, and should be suspected especially following valve replacement.

Keywords

Case reports • Endocarditis • Transcatheter aortic valve implantation • Actinomycosis

ESC curriculum

4.10 Prosthetic valves • 4.11 Endocarditis • 7.5 Cardiac surgery • 9.1 Aortic disease

Learning points

- Actinomycosis is an invasive tumour-like pathogen which requires more than simple blood cultures for diagnosis.
- TAVI-related infective endocarditis is a diagnostic challenge; therefore, a high level of suspicion is needed in patients with after TAVI presenting with unusual symptoms.

Introduction

Transcatheter aortic valve implantation (TAVI)-related endocarditis is a unique entity with special characteristics, and presentation which sometimes differs from a native valve or surgical prosthetic valve

endocarditis. Actinomycosis is a chronic invasive infection caused by *Actinomyces* species, with a tumour-like presentation. Actinomycosis endocarditis has been described, yet considered rare. We present the first reported TAVI-related actinomycosis endocarditis.

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Timeline

Transcatheter aortic valve implantation	
Few weeks after the procedure	Patient developed hoarseness.
Four months after the procedure	Patient was diagnosed with vocal-cord paralysis, developed generalized weakness.
One week later	Admitted to the hospital for evaluation of general weakness. Computed tomography angiography (CTA) demonstrated an aortic pseudoaneurysm.
Two days after admission	Patient developed cardiogenic shock, transferred to the operating room for an emergent surgery included tamponade identification due to aortic dissection, with aortic valve and ascending aorta replacement. Vegetation on TAVI prosthetic valve was sent for cultures.
Within a week from the procedure	Cultures were positive for <i>Actinomyces neuii</i> , ampicillin intravenous therapy was initiated.
Twelve weeks after initiation of therapy	Intravenous antibiotics were replaced by oral amoxicillin.
Four months after the procedure	The patient was rehabilitated and recovered.

Case presentation

A 70-year-old female patient presented to the emergency department with generalized weakness and abnormal laboratory results. Four months before her presentation, she underwent TAVI with SYMETIS SA valve (Boston Scientific Company, Ecublens, Switzerland) due to symptomatic severe aortic stenosis. Her other medical history stands for hyperlipidaemia, essential hypertension, iron-deficiency anaemia, severe kyphoscoliosis, and chronic obstructive lung disease. She was deferred from surgery by the institution's heart team due to her comorbidities.

A few weeks following the TAVI procedure, she was diagnosed with vocal-cord paralysis due to disturbing hoarseness. The paralysis was suspected to be caused by laryngeal nerve compression or infiltration. Shortly after the diagnosis, she presented to the hospital due to general weakness, weight loss, and severe anaemia. On admission, her vital signs were normal. Blood pressure was 130/57 mmHg, heart rate was 88 b.p.m., air room saturation was 98%, and her temperature was measured 37.1°C. On physical examination, heart sounds were normal, without murmurs, breathing sound decreased diffusely, jugular venous pressure was normal, and the thyroid gland was not enlarged. Her laboratory work revealed elevated inflammatory markers with white blood count of 16.12K/ μ L (normal range 4.8–10) with neutrophilia (83%), C-reactive protein was 14.3 mg/dL (normal 0.0–0.5), haemoglobin was 8.5 g/dL (normal range 12–16), creatinine was elevated 1.3 mg/dL (normal 0.5–1.2), and liver enzymes were elevated five times the upper normal limit.

Abdominal sonography was normal. Chest X-ray demonstrated deviation of the trachea and raised the suspicion for mediastinal widening.

Therefore, she underwent a CTA which revealed a 28 mm pseudoaneurysm of the ascending aorta and a small pericardial effusion (Figure 1). Transthoracic echocardiography showed normal functioning prosthetic valve with no vegetations, although a pseudoaneurysm flap was noticed in the aortic root (Figure 2). Two days after her admission, she developed acute dyspnoea with cardiogenic shock. Transoesophageal echocardiography demonstrated a marked thickening of the aortomitral continuity, which was suspected to be an abscess (Figure 3). She was transferred immediately to the operating room for emergent cardiac surgery. After a mid-sternotomy, a tamponade was diagnosed with active bleeding and blood clots in the pericardial cavity. The 2 × 2 cm² pseudoaneurysm was identified protruding from the ascending aorta, with a dissection of the ascending aorta anterior wall from the sinotubular junction. She underwent surgical aortic valve replacement (SAVR) with Perimount 21 mm tissue valve and ascending aorta replacement with interposition graft. A 2 cm vegetation was found on the TAVI prosthetic valve and sent with the involved portion of the aorta for pathology and cultures (Figure 4).

A culture from the vegetation and the ascending aorta pseudoaneurysm revealed an *Actinomyces neuii* infection. Pathology from the ascending aorta was positive for fragments of the aortic wall showing massive transmural necrosis with neutrophilic infiltrates. Repeated blood cultures were negative. Intravenous ampicillin treatment was administered for 12 weeks, followed by 12 months of oral amoxicillin treatment. During the immediate post-surgical period, the patient was haemodynamically stable and both kidney and liver functions gradually improved. However, due to her obstructive lung disease, weaning from ventilation was prolonged and eventually she underwent tracheostomy and was discharged to a rehabilitation centre. She underwent a successful weaning from mechanical ventilation and was discharged home following 25 days of respiratory rehabilitation. Her follow-up echocardiography 6 months following her discharge showed normal functioning prosthetic valve and good left ventricle function.

Discussion

The clinical presentation of actinomycosis, which is caused by members of the genus *Actinomyces*, is characterized by its chronic course and the invasion of multiple tissue planes. Histologically, actinomycosis is characterized by the formation of sinus tracts and masses resembling malignancy which often complicates its diagnosis. It tends to relapse when short courses of antibiotics are used.^{1–3} Actinomycosis typically involves cervicofacial, thoracic, and abdominal regions, but can also cause a variety of other infections, including abscesses, skin infections, osteomyelitis, bacteraemia, central nervous system infections, and many others.^{1–7} Actinomycosis endocarditis has been described, yet it is considered extremely rare.^{5,6}

Organisms belonging to the *Actinomyces* genus are typically aerotolerant gram-positive rods that grow in branching filaments. Isolates are usually catalase and urease negative.⁸ However, thanks to advances in laboratory identification methods, many novel *Actinomyces* species have been reported in recent decades, one of which is *A. neuii*.⁹

Actinomyces neuii has been implicated in several human infections, yet has rarely been reported in the literature. It has been hypothesized that dismissal as a commensal coryneform bacillus may be the reason.¹⁰ Abscesses are the most common manifestation of *A. neuii* infection, with more than half of the reported cases describing abscesses or infected atheromas. Other infectious diseases include skin and ulcer infections, cellulitis, urinary tract infections, prostatitis, endophthalmitis, and bacteraemias.¹¹

Actinomyces neuii endocarditis reported cases are scarce; Yang and Grant¹² presented a patient with an acute *A. neuii* aortic and mitral native valves endocarditis complicated by aortic root abscess and septic cerebral emboli. He was treated successfully with surgery and prolonged antibiotics. Another early report described vegetation on a

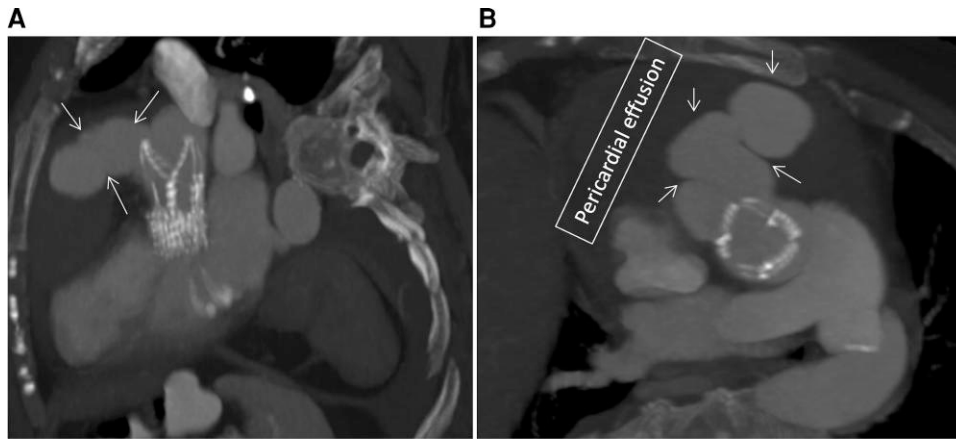


Figure 1 Computed tomography angiography sagittal view (A) and axial view (B) of pseudoaneurysm from the ascending aorta (arrows), in a patient after transcatheter aortic valve implantation. Note the large pericardial effusion.

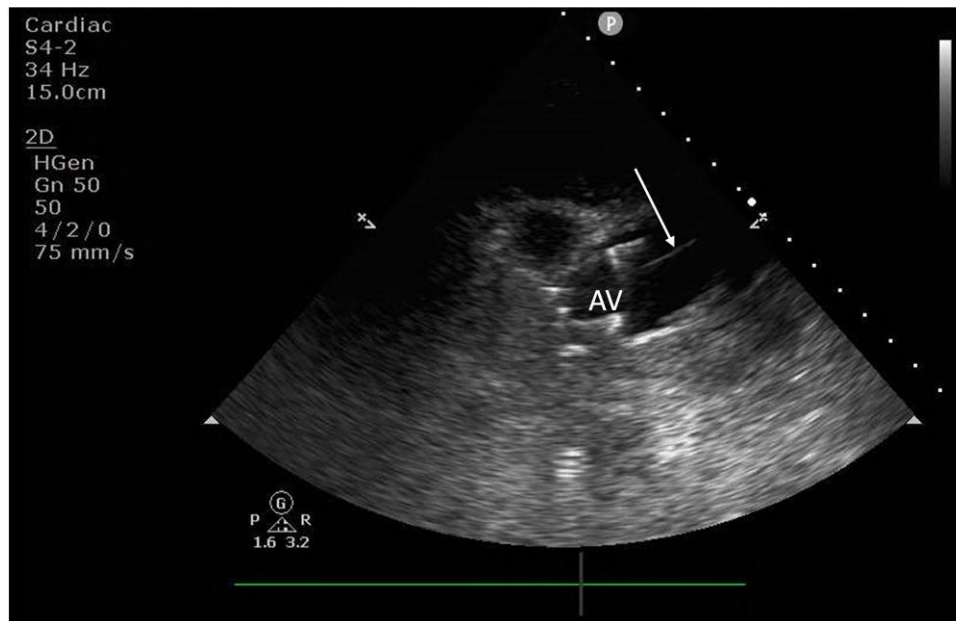


Figure 2 Transthoracic echocardiography showing the aortic valve and proximal ascending aorta, note the pseudoaneurysm flap (white arrow). AV, aortic valve.

bicuspid aortic valve, with a para-valvular abscess attached to the basal septum in a 68-year-old man who presented with fever, treated with surgery and prolonged antibiotic therapy.¹³

A later report described a case of aortic prosthetic valve *A. neuui* endocarditis, in a patient who presented with a 2-month history of dyspnoea, general weakness, and recurrent fever, 4 years after SAVR with 23 mm Medtronic-Hall composite prosthesis and aortic conduit implantation due to thoracic aortic aneurysm and aortic valve insufficiency. This patient was successfully treated by antibiotics only, without surgical intervention.¹⁴ In our case, the clinical presentation was of indolent course at the beginning, which fits actinomycosis natural history. The contamination occurred most probably during the TAVI procedure. The

pseudoaneurysm might be explained by a slow abscess formation and the concomitant emergence of an aortic dissection. After the emergent surgery, long-term antibiotic therapy was recommended, due to the recurrent nature of the pathogen.^{2,3}

The incidence of TAVI-related endocarditis has been reported to be about 1.1–1.8% per patient year and is not significantly different from the incidence of SAVR endocarditis. The diagnosis of TAVI endocarditis is considered more challenging than SAVR endocarditis, due to lower diagnostic yield of echocardiography. Therefore, the Duke criteria has lower diagnostic yield for prosthetic valve endocarditis and many patients are classified as ‘possible’ endocarditis due to negative or equivocal findings.¹⁵

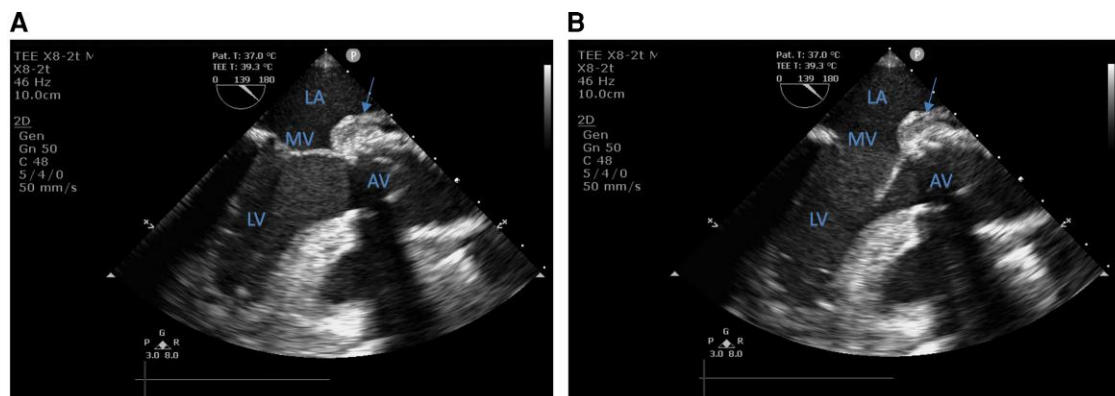


Figure 3 Transoesophageal echocardiography in systole (A) and diastole (B), long axis view. Arrows pointing to the markedly thickened aortomitral continuity, suggesting an abscess formation. MV, mitral valve; AV, aortic valve; LA, left atrium; LV, left ventricle.

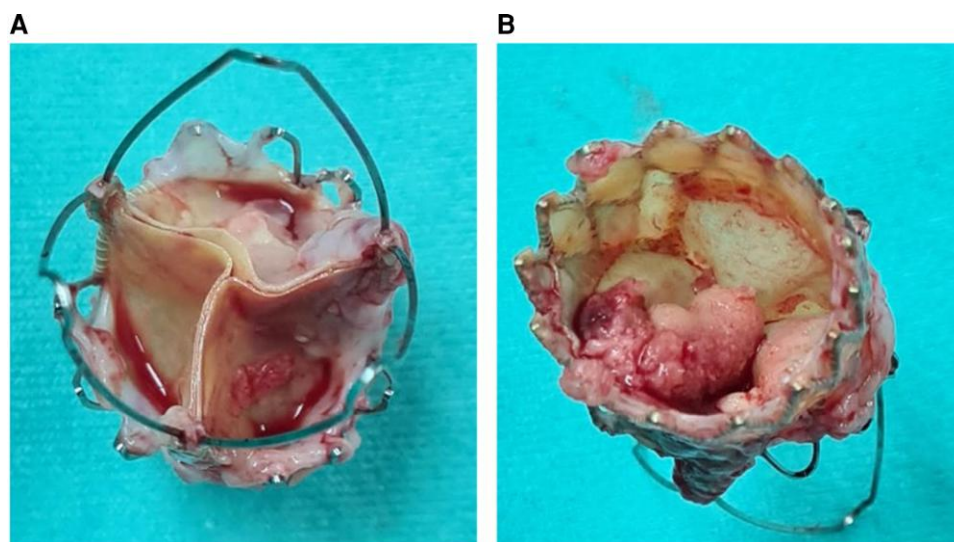


Figure 4 The prosthetic valve (Symetis SA valve; Boston Scientific Company, Ecublens, Switzerland) covered with a vegetation upon its ventricular surface. (A) Aortic view. (B) Left ventricle view.

Nonetheless, unlike SAVR endocarditis, which has been shown to benefit from a multimodality evaluation by cardiac computed tomography (CT) and ^{18}F -fluorodeoxyglucose positron-emission tomography/CT, those modalities have not been sufficiently studied in TAVI endocarditis and the diagnostic value of these techniques is unknown.

The patient presented in this case suffered from prolonged hoarseness before her presentation, and this was her sole symptom. Hoarseness was explained by the bilateral laryngeal nerve compression due to the large ascending aortic pseudoaneurysm, which caused vocal-cord paralysis. She did not present with typical systemic infection symptoms (such as fever, weight loss, or shivering) nor did she suffer from aortic valve dysfunction-related symptoms like dyspnoea, chest pain, and heart failure. The lack of systemic symptoms was probably due to the fact the infection was contained and there was no bacteraemia. Nevertheless, whenever a patient with

prosthetic valve presents with unusual complaints or abnormal laboratory tests (e.g. new onset anaemia, acute kidney injury, etc.), the differential diagnosis of infective endocarditis must be suspected and ruled out.

Conclusion

In conclusion, we presented a patient with an unusual clinical presentation of TAVI endocarditis with a rare pathogen. This case emphasizes the importance of keeping a high level of suspicion for infective endocarditis in every patient with prosthetic valve. It is important to remember that unusual pathogens, especially actinomycosis, often results in unique presentation, and may necessitate a more comprehensive diagnosis. Finally, to our knowledge, this is the first actinomycosis endocarditis in TAVI valve reported to date.

Lead author biography



Dr Chen Gurevitz has specialized in internal medicine and is a cardiology fellow in Rabin Medical Center, Israel. She is the head of Lipids Clinic in the cardiology department.

Supplementary material

[Supplementary material](#) is available at *European Heart Journal—Case Reports* online.

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Slide sets: A fully edited slide set detailing these cases and suitable for local presentation is available online as [Supplementary data](#).

Consent: The patient have signed a consent statement for the publication of this case report in accordance with COPE guidelines.

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